

“A Cystic Neoplasm in Disguise”

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ABSTRACT

Odontogenic lesions are diverse pathologies of jaw representing a wide variety of lesions which develop from remnants of tissue associated with odontogenesis. It includes various cysts which are commonly encountered in dental practise and tumours which are relatively uncommon accounting for less than 1%. These can develop at any age and any site of the jaws. They may remain occult or can be aggressive. We report a unique case of extensive multilocular lesion of the mandible with cortical bone expansion and root resorption in a 40 year old female. Although the closest differential diagnosis was ameloblastoma, the histopathological examination revealed a cystic lesion with diverse features which was finally concluded as Keratocystic odontogenic tumour. The present case is unusual in terms of its site of occurrence, presentation and histopathology emphasizing the importance of incisional biopsies from multiple sites in case of extensively large lesions and meticulous histopathological examination of the entire lesion for establishing a final diagnosis.

Keywords: Keratocystic Odontogenic Tumour, Reduced Enamel Epithelium, Anterior Mandible.

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Introduction

Odontogenic pathologies are a group of site specific lesions occurring in jaw bones unless metastasized to other body sites. These pathologies vary in nature from hamartomatous proliferations to malignant neoplasms with metastatic potential. The source of origin being tissue associated with odontogenesis[1], (cell rests of serres, epithelial cell rests of malassez, reduced enamel epithelium and the ectomesenchymal element). They can remain restricted to a localized region or can extensively involve the affected jaw. Mandible is more commonly involved with mandible to maxilla ratio of 4.5:1[2]. An understanding of biologic behavior is of fundamental importance to the diagnosis and treatment planning which should be achieved by correlating historical, clinical, radiographic and histologic features[3].

Here we present a case of extensive lesion of mandible, pre-



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sented as a multilocular lesion with buccal and lingual cortical bone expansion.

Case Report

A 40 year old female reported with a chief complaint of slowly enlarging mandible since 10 years to our department. Patient was moderately built and well nourished with all the vital parameters in normal range. There was no significant medical history. Patient had swelling from one side of the mandibular body region to the other, i.e. across the midline but more in the symphyseal region with expansion in transverse and vertical dimension.



Fig.1 Preoperative Picture

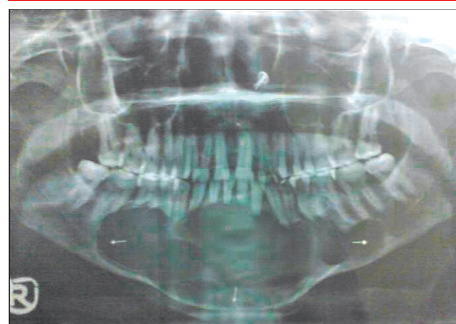


Fig.2 Digital OPG showing the extent of lesion.

Intraorally bony expansion was seen on buccal as well as lingual side from 37 to 47 region [Fig.1]. It was firm and non tender on palpation. Aspiration revealed brownish-orange colour fluid. Radiographic examination revealed a multilocular radiolucent lesion with well-defined borders extending from 37 to 47 regions with thinning of inferior border of mandible and root resorption of the associated teeth [Fig.2]. A provisional diagnosis of ameloblastoma was established. All the necessary investigations were performed before taking the patient for general anaesthesia. Intraoperatively visor flap was raised from one angle of mandible to the other and lesion was carefully separated from the soft tissue margins. Facial artery and vein were ligated on both the sides along with preservation of marginal mandibular branch

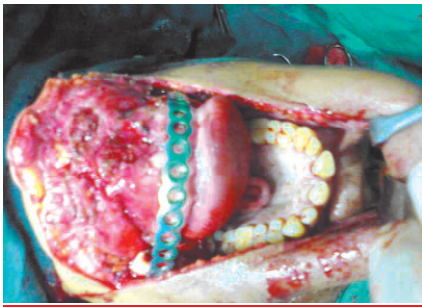


Fig.3 Reconstruction Plate in situ



Fig.4 Resected Mass

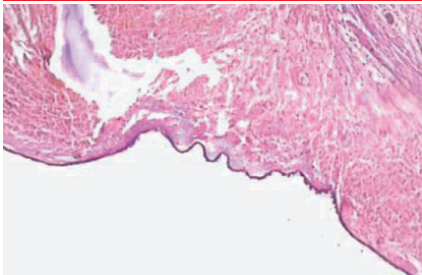


Fig.5 Cystic Lining of non-keratinized squamous epithelium resembling REE

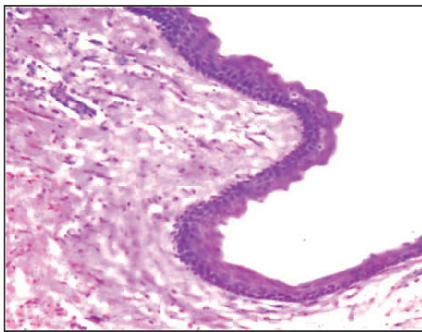


Fig.6 Uniform Lining of Thickness of 6-8 Cell layers

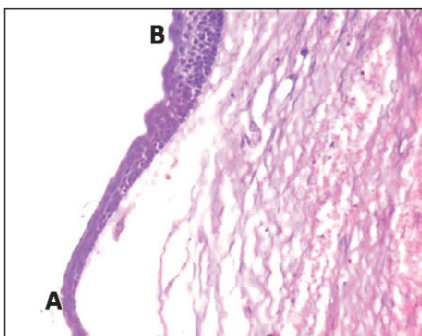


Fig.7 Transition of REE Like(A) Lining to KCOT Lining(B)

of facial nerve. Separation from the lingual side was done taking care that submandibular salivary duct and lingual nerve were not damaged. Surgical resection of the mandible from 37 to 47 region was performed with 1.5 cm of healthy bone margins. Reconstruction was done using 2.7 mm reconstruction plate and closure was done in layers [Fig.3]. Gross examination of the excised specimen showed completely hollowed-out cortical plates and perforation of buccal cortical plate [Fig.4]. Extensive histopathological examination of the excised specimen revealed a cystic space lined predominantly by thin, non-keratinized squamous epithelium resembling reduced enamel epithelium (REE) [Fig.5] at 4X Magnification. In areas the lining was characteristically of uniform 6 to 8 cell layer thickness with pallasided basal cuboidal to columnar cells containing reversely polar, hyperchromatic nuclei. The surface was corrugated parakeratinized [Fig. 6] at 40X magnification. The REE like epithelium was in direct continuity with the typical Keratocystic odontogenic tumour like lining [Fig.7] at 10X magnification. A final diagnosis

of Keratocystic odontogenic tumour was established.

Postoperative healing was uneventful and movements of reconstructed mandible were satisfactory. Patient is scheduled for complete reconstruction with free fibula and microvascular anastomosis after the observation of a disease free state of 1 year.

Discussion

Multilocular radiolucencies of the mandible are not uncommon, the commonest lesion being ameloblastoma accounting for 72 % of these tumours[2]. They are produced due to multiple, adjacent often coalescing and overlapping pathologic compartments in the bone. The other differential diagnosis includes odontogenic keratocyst, dentigerous cyst, radicular cyst, central giant cell granuloma, fibro-osseous lesions[4]. KCOT usually are unilateral lesion growing along the bone marrow without causing expansion of buccal and lingual cortical plates[4]. In the present case, the expansion, thinning and perforation of cortical plates were the unusual findings which mislead us to a provisional diagnosis of ameloblastoma. A similar case of massive KCOT was reported in an 11 year old female involving from 36 to 46 region but without perforation of cortical plates[5].

KCOT is a distinctive cystic lesion characterized by different growth mechanism and biologic behavior than the other odontogenic cysts[6]. Debate still exists regarding its cyst vs cystic neoplasm categorization. However, the latest WHO classification 2005 of odontogenic cysts and tumors has introduced the term keratocystic odontogenic tumor (KCOT) and classified it under the category of benign tumours arising from odontogenic epithelium without ectomesenchyme[7].

The KCOT is estimated to make up 8% to 11% of all odontogenic cysts[8]. The commonest site is mandible, particularly the posterior body and ramus region. It may also occur in other rare sites like anterior mandible and maxilla[6,9]. The peak frequency of occurrence is in second and third decade of life with slight male predilection[9]. Although the age of occurrence is in concordance with earlier reports [5,6,7] but the location was unusual in the present case.

The histopathology in the present case showed reduced enamel epithelium like lining in direct continuity with typical KCOT lining [Fig.5]. Pathologists should be especially cautious and critical when recognizing and interpreting the changes within the epithelium of a dentigerous cyst while differentiating it from KCOT and unicystic ameloblastoma and also for any possible cancerization[10].

Yoon et al.[11] in 2006 reported an unusual odontogenic cyst presenting as a dentigerous cyst with varied histological features like ciliated, glandular and squamous epithelium with keratinization. Multipotentiality of odontogenic epithelium is well established[11] substantiating a possible pathogenic mechanism of transition of one type of epithelium to the other. A final diagnosis of keratocystic odontogenic tumour was arrived at as it would

define the biologic behavior in the present case [Fig. 4].

The various surgical treatment methods for KCOT can be categorized as conservative or aggressive[12]. The type of treatment chosen depends on several factors like age of the patient, location and size of the lesion and also whether it is primary or recurrent KCOT [5]. Occasionally, a locally aggressive odontogenic keratocyst cannot be controlled without local resection[6]as was done in present case. The overall prognosis for most KCOT is good. KCOTs have a high recurrence rate, ranging between 25% and 60%[1]. Recurrence rate is relatively low with aggressive treatment. A systematic review done on KCOT reported 0% recurrence after resection[13].

The possibility of malignant transformation, high recurrence rate and association with nevoid basal cell carcinoma syndrome makes KCOT of considerable importance[13] and thus warranting a regular and vigilant follow-up.

Conclusion

The present case is unique in terms of its site of occurrence, clinical and histopathological presentation. With this we would like to conclude that: All massive aggressive multilocular lesions of mandible are not always ameloblastoma. In case of huge lesions, incisional biopsy from different sites is advocated. Multipotentiality of odontogenic epithelium necessitates thorough histopathological examination of the entire specimen.

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